

## BRANCHIOGENETIC CYSTS\*

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**B**RANCHIOGENETIC, or branchial cysts, occur frequently enough to occasion a review of their characteristic features and the problems relating to their successful care.

We may classify the varieties of cysts in the neck as those lying in the midline, such as thyroglossal, dermoid and thyroïd cysts, and those situated laterally, namely, branchiogenetic, hygroma, and parasitic cysts. Such possible entities as tuberculous abscesses, esophageal diverticula, and aneurysms are not classified as cysts, but they assume tremendous proportions when a differential diagnosis must be made.

This discussion deals with the diagnosis and treatment of thirty branchiogenetic cysts. Four of the patients so affected have been operated by me, the other twenty-six having been treated during the last decade by the staffs of the Los Angeles General Hospital and the St. Vincent's Hospital of Los Angeles.

## EMBRYOLOGY

A review of the embryologic disposal of the fetal branchial apparatus is important for the comprehension of our subject. Rathke, in 1835, first described branchial clefts in the mammalian embryo. Twenty-four years later Rosen placed lateral cervical cysts on an embryologic basis as being of

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Read before the General Surgery Section of the California Medical Association at the sixty-sixth annual session, Del Monte, May 2-6, 1937.

branchial origin. Not until 1912, however, when Wenglowski<sup>1</sup> published his thesis, was there any definite progress made in the study of the embryologic origin of branchiogenetic cysts. His theory ascribed their origin to remnants of the embryologic thymic duct, and indirectly to the branchial apparatus. Present authorities agree with this theory.

The branchial system appears in the third or fourth week of intra-uterine life, and is completely gone at the eighth week. There are five or six branchial arches which are separated by depressions, otherwise known as branchial grooves. The inner view of the pharynx shows these depressions, which are spoken of as pharyngeal pouches. In the gill-bearing animals the ectoderm and endoderm disappear in places and gill slits are formed.

Embryologists are quite agreed regarding the development of the branchial structure. The first arch divides, the superior portion forming the upper lip and part of the maxilla and cheek. The inferior portion forms the lower lip, a part of the mandible, the tongue and muscles of mastication. The first cleft forms the auditory canal and the lobe of the ear. The first pharyngeal pouch gives rise to the eustachian tube and the tympanic cavity. The tympanum is derived from the first gill membrane.

From the second branchial arch are derived the lesser cornu of the hyoid bone, the styloid process, and the stylohyoid ligament. The muscles of the base of the tongue, in its anterior portion as well as the arcus palatoglossus, are of this same origin. The tonsil and sinus tonsillaris develop from the second pharyngeal pouch.

From the third arch are derived the greater cornu of the hyoid bone, the stylopharyngeus muscle, and a portion of the muscles of the soft palate. From the third pharyngeal pouch the thymus has its origin, and from remnants of this structure bran-

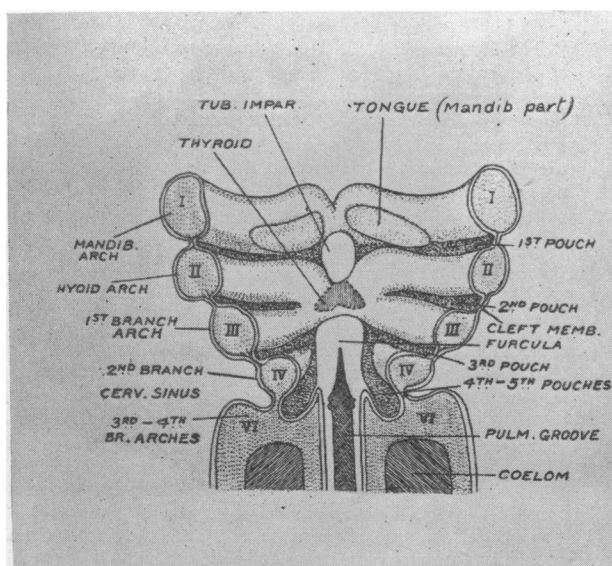


Fig. 1

Fig. 1.—Showing the floor of the pharynx of a fifth week human embryo; arches shown with Roman numerals. (After Keith.)

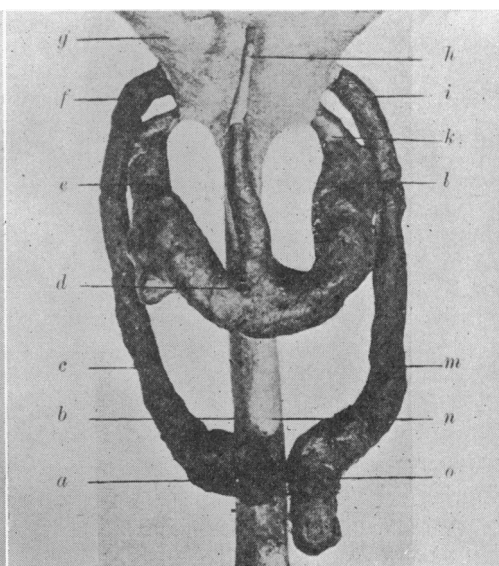


Fig. 2

Fig. 2.—Fourteen-millimeter embryo, showing development of pharynx, esophagus and trachea and related structures: a, thymus; b, esophagus; c, thymic duct; d, midthyroid lobe; e, lateral thyroid lobe; f, thymus duct; g, pharynx; h, thyroglossal tract; i, thymus tract; k, duct of lateral thyroid lobe; n, trachea; o, thymus. (After Wenglowski.)

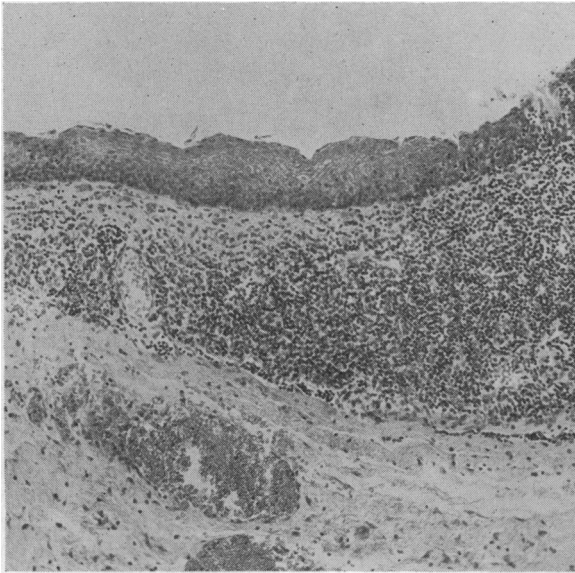


Fig. 3

Fig. 3. Medium-power photomicrograph through the wall of a branchial cleft cyst, showing a stratified squamous epithelial lining with lymphoid tissue in the connective tissue corium.

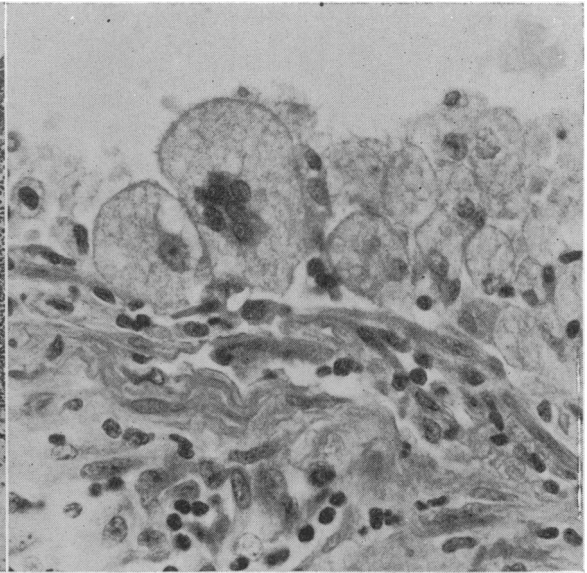


Fig. 4

Fig. 4.—High-power photomicrograph of branchial cleft cyst, showing degeneration of the epithelium and phagocytosis of the lipoidal debris by macrophages.

chiogenetic cysts are formed. The fourth pharyngeal pouch gives rise to the lobes of the thyroid gland. The remaining fourth, fifth, and sixth arches lose their identity, but make up the soft parts in the tissues adjacent to the greater cornu of the hyoid bone.

#### WENGLOWSKI'S THEORY

The preceding reference to the thymus structure recalls Wenglowski's theory that cervical cysts and fistulae must have their origin in the thymus apparatus, which appears and descends laterally as a tubular structure from the third pharyngeal pouch in the 6.5 millimeters embryo to the lower border of the lobes of the thyroid. Here it becomes a gland-like organ which, at a later time, descends under the sternum and becomes the thymus gland. The trailing tubular structures normally disappear. Wenglowski stated that cervical cysts and fistulae must arise in remnants, as they are always located in the position and extent of the fetal thymic structure. To this most authorities now agree. He emphasized further that, since the main branchial apparatus does not extend below the hyoid bone, fistulae and cysts of branchial origin could not be found below this level. Since fistulae and cysts do occur below this level, the thymus anlage must be their origin.

We infrequently encounter an internal fistulous opening, in the suprathyroidal fossa derived from the second pharyngeal pouch. That it is a remnant of the upper portion of the thymic duct is contradictory to the Wenglowski theory. An instance is reported where a patient could inflate a branchiogenetic cyst via the pharynx.

We, therefore, are committed to Wenglowski's theory in the explanation of the genesis of branchiogenetic cysts. The histologic picture conforms to misplaced epithelium. This varies from the stratified squamous type of epithelium to the transitional

variety in the cysts high in the neck. In some instances the lining epithelium is columnar, often ciliated, and in these instances the cysts are found in a low position in the neck.

The wall of the cyst is composed mostly of dense fibrous tissue with lymphoid follicles scattered throughout. With bacteria present and usually the result of upper respiratory infection, the character of the retained material, and the reaction of the epithelial and lymphoid structures become altered.<sup>3</sup>

#### HYGROMA

Hygroma,<sup>4</sup> or lymphocoele, is the most important cyst to be differentiated from the branchiogenetic type, its embryology being also important. It is of lymphatic origin, and arises from the jugular sinus seen in the third month of fetal life. The lymphatic

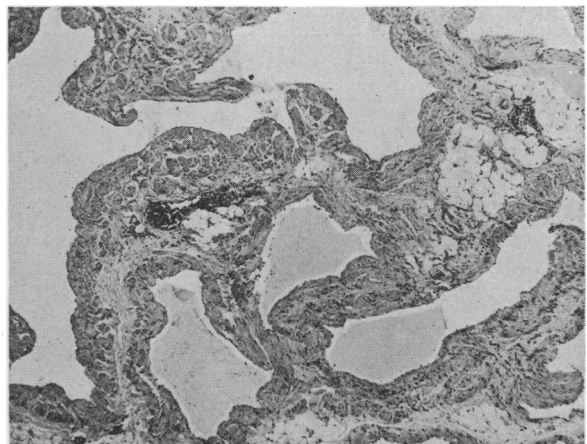


Fig. 5.—Low-power photomicrograph of cystic lymphangioma of the neck, showing dilated lymphatic spaces with well-developed muscle walls and endothelial lining. Occasional nodules of lymphocytes are noted in connective tissue, and coagulated lymph is seen in some of the cystic spaces.

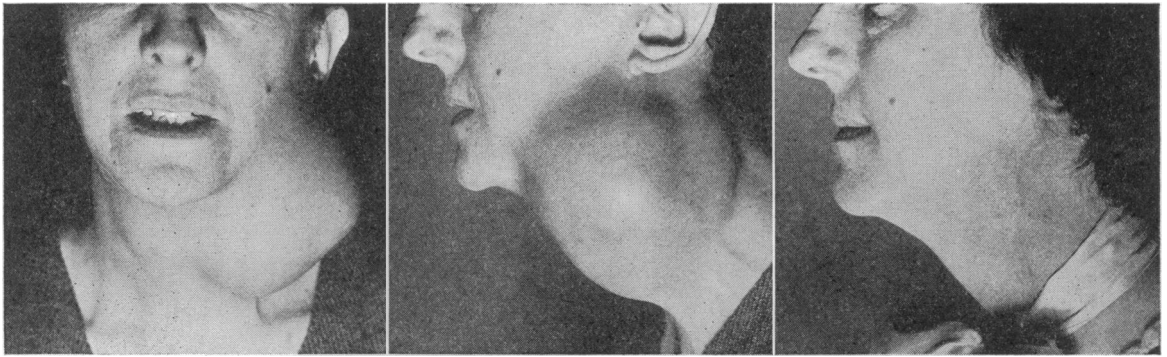


Fig. 6

Fig. 7

Fig. 8

Fig. 6 (V. S., age 39).—Branchiogenic cyst of eight years' duration. Extremely large and infected. Removed with vertical incision. Local anesthesia. Front view.

Fig. 7.—Side view of the same patient. Note location and lobulation of the cyst.

Fig. 8.—After six months. Vertical incision has produced a minimal scar.

structures are well developed in 30 millimeters embryo. They are ordinarily obliterated by connective tissue, and eventually form lymph follicles and gland nodes which originate from the endothelium lining the primary sacs. Structural faults may occur with failure to obliterate all or part of these sacs, which may develop cystic tumors of the neck, from the level of the styloid down to the axillary space. A recent patient was operated by us in whom the cyst was located in the right cheek and jaw region. Such cysts are commonly found in children in the posterior triangle and in the right side of the neck.

#### DIAGNOSIS

The diagnosis of branchiogenic cysts can be made on finding a cystic tumor in the neck, laterally located and lined with epithelium, the wall consisting of dense fibrous tissue with lymph follicles scattered throughout. The content of these cysts is the product of epithelial metabolism, and when infection is present the character of the aspirated fluid is altered from the normal by the extent, type, and virulence of the infecting organism. The tumor is cystic to palpation and in many instances there is the sense of lobulation. Tenderness is absent, but it may exist in varying degree when infection is present. Diagnosis can be made by aspiration and careful microscopic examination of the retained fluid. Caution must be exercised against the introduction of bacteria when aspiration is made.

Because hygroma is of endothelial origin and contain lymph, aspiration and examination of the fluid make the diagnosis accurate. A cross section shows a sac lined with endothelium, containing a cobweb-like structure filled with lymph. The diagnosis of tuberculous abscess must be made on the infiltrating character of the tumor, and the absence of epithelial debris and lymph in the aspirated specimen.

#### TREATMENT

The treatment of branchiogenic cysts requires complete surgical removal. The incision is made over the cyst, parallel to the edge of the sternocleidomastoid muscle. Careful dissection must be made, as the wall of the cyst can easily be ruptured.

The carotid vessels should be identified and injury avoided. In the superior medical border of the cyst the surgeon must exercise care to remove an incomplete tract or a possible fistula communicating with the supratonsillar fossa. Failure to recognize this structure may lead to a recurrence of the cyst or to a fistulous tract requiring further surgical procedures. Evacuating aspirations may be employed as a temporizing measure. The danger of introducing infection and the improbability of cure by this means should place this procedure in the discard.

Hygroma, on the other hand, is usually a difficult surgical procedure because of the tedious dissection of the many processes which may widely penetrate the adjacent structures. Rapid recurrence follows incomplete removal. One of our patients was operated upon twice by us, but there has been no further recurrence. Destruction with a sclerosing solution may be of great value, though I can find no report of the results of this method of treatment for hygroma.

Table 1, summarizing thirty instances of branchiogenic cysts, reveals that the cyst is located on either side of the neck in almost equal proportions. The same distribution is true in respect to the gender of the patients. There were five recurrences (16 per cent), resulting from incomplete removal. One patient developed branchiogenic carcinoma. It is interesting to note that in all instances the epithelium was of the stratified squamous variety, indicating high location of the tumor in the neck. In one patient there had been many aspirations with infection of cystic contents before total removal was done. It is interesting to note that one patient carried his cyst for nineteen years, and was operated upon three times during that period. At the last operation the insertion of a tracheotomy tube was necessary. Because of incomplete histories it is impossible to state the average duration of the existence of the cyst. These cysts occurred in middle life in most instances. In one patient the wound is still draining one year subsequent to operation, indicating that the cyst has not been completely removed. Local anesthesia was used in only one operation.

TABLE 1.—Summary of Thirty Branchiogenetic Cysts—16 Per Cent Recurrence, 3⅓ Per Cent Malignant Change—Sex and Position Equal—Pathology Same in Each Instance—Duration Ranges from Three Weeks to Nineteen Years in Second, Third and Fourth Decades of Life

Name	Sex	Age	Side	Duration	Anes- thetic	Pathology	Miscellaneous
H. B.	M	25	R	3 months	?	Strat. sq. epi. and lymph nodules	
R. R.	M	27	L	?	Ethylene	"	
P. K.	F	36	L	3 weeks	Ether	"	
E. H.	M	17	R	2 years	Ether	"	
J. C.	M	17	L	1 year	Ether	"	
V. S.	F	39	L	8 years	Local	"	
M. H.	F	49	R	2 years	Ether	"	
O. M.	F	43	L	18 months	Ethylene		
M. C.	F	24	L	?	Ethylene		
H. K.	M	3	R	1 year	Ether		
G. H.	M	?	L	1 year	Local	"	
Z. G.	F	22	L	2 years	Ether	"	
T. G.	F	38	R	9 months	Ether	"	
P. G.	F	36	R	4 months	N <sub>2</sub> O	"	
W. B.	M	51	R	5 months	Ether	"	
J. N.	M	29	L	2 months	Ether	"	
L. B.	M	28	R	3 months	Ether	"	
R. D.	M	30	R	4 years	N <sub>2</sub> O	"	
J. M.	M	46	R	9 years	N <sub>2</sub> O	"	
R. B.	F	44	L	?	Ether	"	
M. V.	F	22	L	?	Local	"	
G. B.	M	33	L	?	Ether	"	
W. H.	M	20	R	14 months	?	"	Two operations
T. P.	M	20	R	2 years	N <sub>2</sub> O	"	Two operations
R. S.	F	19	R	19 years	?	"	Three operations
V. M.	F	21	L	5 years	N <sub>2</sub> O	"	Three operations
C. C.	M	28	L	7 years	Ether	"	Aspirated many times
W. K.	F	28	R	6 months	Ether	"	Still draining after one year
J. A.	M	54	R	1 week	Ether	"	Thyroidectomy 3 weeks later
T. P.	M	43	R	?	?	"	Branchiogenetic cancer

SUMMARY

Branchiogenetic cysts occur in either side of the neck in equal proportions in both male and female. These cysts, lined with epithelium, arise from remnants of the thymus anlage and contain débris of epithelial metabolism. The walls are composed of connective tissue with lymph follicles scattered throughout. Complete surgical removal is mandatory. Hygroma, or lymphocoele, which occurs usually in childhood, approaches more nearly the characteristics of the branchiogenetic cysts. Hygroma is readily diagnosed, because of the endothelial components of the tumor and the finding by aspiration of coagulable lymph.

1930 Wilshire Boulevard.

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DISCUSSION

ARTHUR E. SMITH, M.D. (1930 Wilshire Boulevard, Los Angeles).—Doctor Larson has brought to our attention a most interesting subject, and one that has intrigued me for many years. The diagnosis and treatment of cystic tumors of the neck present a problem with which the general practitioner is not quite so familiar, due to their comparative rareness.

A report of thirty instances of bona fide branchiogenetic cysts operated upon, with recurrence of 16 2/3 per cent, brings to our mind the importance of a thorough knowledge in the surgical treatment of this type of condition. One case in which he reported the occurrence of branchiogenetic cancer brings out a fact which must be borne in mind at all times: in patients with cysts of long standing we must always consider the possibility of malignancy, and always assure ourselves that it is not present before discharging the patient as cured.

From the standpoint of treatment the differential diagnosis between branchiogenetic cysts and hygroma is most important. Whereas branchiogenetic cysts are removed with relative ease and with comparatively little shock, the prognosis in hygroma must always be guarded due to the

nature of the cyst and the difficulty attending its removal. The diagnosis can usually be made by simple aspiration and examination of the contents under the microscope, since branchiogenetic cysts contain the products of epithelial metabolism, while hygromas contain the products of endothelial metabolism.

Whether these cysts and fistulae are developed from the remnants of the primitive thymic ducts, or whether they have other sources of etiology, is a problem that we cannot take up too seriously at this time and one in which we should not form hasty conclusions. Most authorities agree that Winklowski's ideas fit easily into the picture of these cysts and are more plausible than those offered by any other theory. However, for the present this need have no bearing on the treatment, since radical extirpation is mandatory in all cases.

The essayist has called attention to the necessity of being certain that the cyst in its entirety be removed. We would like to further emphasize this point and further admonish the surgeon to remove any possible fistulous tract that may lead to the supratonsillar fossa. Unless these factors are borne in mind, and unless the surgeon employs careful dissection, the result of the operation will be failure in 100 per cent of the cases. In the series reported by Doctor Larson there were recurrences which is entirely too high. If the treatment of this condition is carried out in a thoroughly painstaking manner, recurrences are very rare.

In the instance where we removed the extraordinary large cyst, local anesthesia was used. We were both amazed at the efficiency with which this work could be done without the use of a general anesthetic. The infiltration of novocain assisted greatly in the dissection of the cyst wall from the neighboring structures, and the capillary oozing was considerably diminished by the small amount of adrenalin in the solution. The patient responded remarkably well, showing no signs whatsoever of shock. It is our intention to use local anesthesia exclusively on these patients in the future.

L. R. CHANDLER, M.D. (Stanford University Medical School, San Francisco).—It has been a pleasure to hear Doctor Larson's presentation and see his "movie" film showing the removal of branchiogenetic cysts. His review of the embryology of the branchial apparatus is thorough, but I do not believe all workers are agreed as to the exact and detailed origin of these cysts. Many branchiogenetic cysts are located near the external ear, or are in front of the tragus, and undoubtedly come from the first rather than the third cleft or pouch.

In reviewing our experiences with branchiogenetic cysts at the Stanford clinics, I find our cases very similar to those reported by Doctor Larson. It is interesting to note that the great majority of these cysts make their presence known after puberty, although they are congenital in their origin. Complete surgical removal is always indicated. These cysts should not merely be incised and drained, as it usually results in infection and a persistent sinus which is more difficult to remove than a cyst. Unless there is some particular contraindication in a given case, I prefer a transverse or slightly oblique line of incision in the neck rather than the vertical incision demonstrated in the film.

JOHN HOMER WOOLSEY, M.D. (Woodland Clinic).—Attention to the embryology of branchiogenetic cysts cannot be stressed too much, for upon this depends the location, the possible points of extension and, therefore, the recognition of the possibility as the probable diagnosis and, finally, the treatment. Doctor Larson's reference to the work of Winklowski is important, for it is based on studies of the embryology in the human, whereas previous studies taught in medicine were based on studies on the guinea pig and rat, and are not exactly comparable. The location and extension of cysts in the persisting thymus-duct tract have, in my experience, run entirely regular as to relation to various structures such as the carotids, the hypoglossal and the glossopharyngeal nerves, and the styloid processes. A recognition of the location and possible extent of a persisting thymus-duct tract will prevent the altogether too frequent removal of the external orifice and some local fat, as is so frequently done with a preoperative diagnosis of sebaceous cyst.

The use of sclerosing solution for these cysts, and for the persisting thymus-duct tract, has been made. I mention it, but to condemn it. Experience demonstrates that the eradication of all the living epithelial cells by this method will not occur, and there results, due to a fibrosis here and a remaining epithelial cell there, recurrent multiple cysts—more difficult than ever to remove.

Surgery, with excision, is the only method to be employed. Aids are the study preoperatively by x-rays, having the tract and cyst, if there is clinical evidence of some connection with the surface either internally or externally, injected with an opaque substance; a preoperative staining of the tract with dye; or, better, an injection of the tract with a colored paraffin mixture (an oil dye as used by painters, in contrast to an aqueous dye, is necessary for this), with such a melting point that it can be injected while in the fluid state and then harden sufficiently to give a solid form to the tract. The latter aids greatly in the surgical dissection.

## THE LURE OF MEDICAL HISTORY†

### SARRÍA'S TREATISE ON THE CESAREAN OPERATION, 1830\*

By SHERBURNE F. COOK, Ph.D.

Berkeley

III‡

VI. *The Actual Practice of the Cesarean Operation.*—One must have already at hand some tepid water for baptism, a scalpel (a kind of knife used by surgeons), with which it would be a good thing for every priest to be provided. In default of such a knife a shaving razor will serve, and if this is lacking, a well-sharpened penknife. Also one should have at hand some good wine and some aguardiente. In addition, there should be prepared and ready swaddling clothes for the infant, and some cloths to absorb and remove the blood and fluids which flow from the incisions which have to be made. All this being in readiness (for this purpose and all the following, one or two women, who are not delicate, but strong and nonpregnant, may assist), the corpse is grasped and arranged on a table or bed, in a supine position, that is, with the mouth upward. Below the waist is placed a pillow or a pile of clothing so as to raise the abdomen higher. When the body has been placed in this position it is covered with a sheet from the breast up, and the pubic region down, leaving only the abdomen exposed. The act is then commended to

†A Twenty-Five Years Ago column, made up of excerpts from the official journal of the California Medical Association of twenty-five years ago, is printed in each issue of CALIFORNIA AND WESTERN MEDICINE. The column is one of the regular features of the Miscellany department, and its page number will be found on the front cover.

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Of interest, in reference to these experiences related in the articles on "Sarría's Treatise on the Cesarean Operation, 1830," is the Associated News dispatch of September 2, 1937, which follows:

#### TWINS BORN WHEN MOTHER DIES GAIN

Infant twins—a boy and a girl—gained strength in Kingsville Texas, although their mother had died before they were born.

The mother, Mrs. George Conde, forty-one, wife of a tenant farmer, was brought to the Kleberg County Hospital on Monday suffering from a heart attack.

As the case grew worse Dr. D. A. Harrison was called. Dr. C. C. Carrithers accompanied him. "When she died," Doctor Harrison said, "everything was ready, so we just went ahead."

The boy weighed 4 pounds 12 ounces and the girl 5 pounds 8 ounces.

† Part I of this paper appeared in the August issue, on page 107; Part II, in the September issue, on page 187. Part III is the concluding paper.